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An extremely rare case of thyroid malignancy from the non-Alpine region: Angiosarcoma

Ömer Bayır[a,*], Demet Yılmazer[b], Reyhan Ersoy[c], Yasemin Akca[b], Güleser Saylam[a], Ünsal Han[b], Ali Özdek[a], Bekir Çakır[c], Mehmet Hakan Korkmaz[d]

[a] Ministry of Health, Dışkapı Yıldırım Beyazıt Research and Training Hospital, Department of Otolaryngology and Head & Neck Surgery, Ankara, Turkey
[b] Ministry of Health, Dışkapı Yıldırım Beyazıt Research and Training Hospital, Department of Pathology, Ankara, Turkey
[c] Atatürk Research and Training Hospital, Department of Endocrinology, Ankara, Turkey
[d] Yıldırım Beyazıt University, Medical Faculty, Department of Otolaryngology and Head & Neck Surgery, Ankara, Turkey

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ABSTRACT

INTRODUCTION: Thyroid angiosarcoma is a rather rare malignancy featuring a poor prognosis, and which may interfere with other aggressive thyroid tumors; it is usually seen in the Alpine region.
CASE PRESENTATION: A 74-year-old male was referred to our center with complaints of progressive neck swelling and dyspnea. He had multiple nodules featuring cystic degeneration and calcifications in the thyroid gland, together with multiple lymphadenopathies of the neck region. Fine-needle aspiration cytology (FNAC) confirmed the presence of anaplastic carcinoma. A total thyroidectomy was performed. During the postoperative period, multiple drainage were performed for recurrent hematomas, but hematoma development could not be prevented. On postoperative day 7, the patient died due to multiple-system failure. Histopathological investigation of the thyroidectomy specimen indicated that the lesion was an angiosarcoma.
DISCUSSION: The cytological diagnosis of thyroid angiosarcoma is quite difficult. Extracapsular invasion and distant organ metastasis during surgery are known as strong and negative prognostic factors for thyroid angiosarcoma. Treatment is quite difficult, since this tumor is locally aggressive, destructive, and features a high recurrence rate. In this case, since extracapsular invasion, as well as lymph node and lung metastasis were present at the time of surgery; the expected survival time was quite short.
CONCLUSION: This case shows that during differential diagnosis, patients initially diagnosed with anaplastic carcinoma via FNAC may actually present with angiosarcoma. It may be helpful to review the treatment modalities for this cancer type, which has a rather poor prognosis and features severe bleeding, as well as local and distant metastasis.

1. Introduction

Thyroid angiosarcomas are rare and very aggressive malignancies [1]. Most thyroid angiosarcoma cases are reported from the Alpine region; only a few cases are reported from the areas beyond this region. This cancer, which is reported in advanced ages, generally develops as a multinodular goiter [2,3]. This tumor originates from the vascular structures of the thyroid gland and is recognized histopathologically by its atypical endothelial cells; it is also associated with the presence of vascular canals microscopically. In addition to its resemblance to anaplastic cancer based on the presence of atypical cells, its aggressive and destructive nature can result in misdiagnosis [1]. Patients commonly present with compression symptoms including a lesion on the neck, dyspnea, hoarseness, and dysphagia due to the rapid growth of the tumor. In many cases, systemic metastasis is present at the time of diagnosis and thyroid angiosarcoma commonly has a tendency of metastasizing to local regional lymph nodes and the lungs [2–4]. In this paper, a 74-year-old male patient who underwent fine-needle aspiration cytology (FNAC), who was initially diagnosed with anaplastic carcinoma, and who was later diagnosed with angiosarcoma following total thyroidectomy is presented. Appropriate treatment modalities are subsequently discussed.

2. Case presentation

A 74-year-old male patient complaining of dysphagia, moderate dyspnea, hoarseness, and neck pain for 1 month was admitted to our tertiary center. Neck ultrasound (US) revealed a conglomerated isoechoic nodule that almost totally filled the right lobe; the nodule

* Corresponding author at: İrfan Baştuğ Cd. S.B. Dışkapı Yıldırım Beyazıt Eğitim ve Araştırma Hastanesi, Kulak Burun Boğaz Kliniği, Dışkapı, Altındağ, Ankara,Turkey. Fax: +90 312 318 66 90.
E-mail address: bayiromer@hotmail.com (Ö. Bayır).

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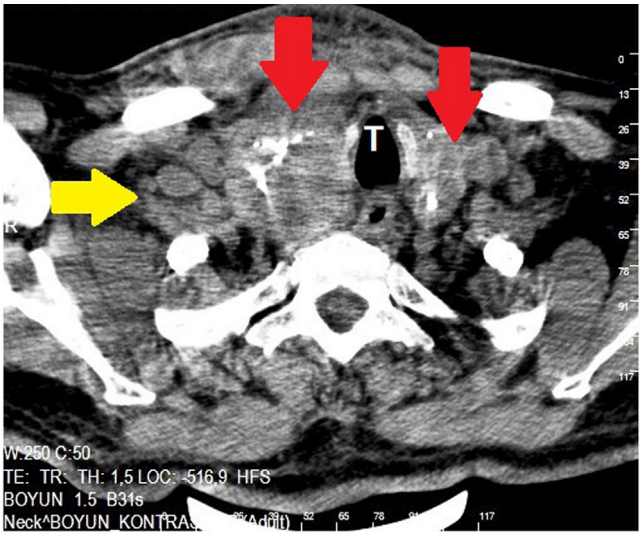

Fig. 1. Preoperative neck CT images of the patient. The trachea is seen to be deviated to the left. Red arrow: thyroid gland featuring calcifications and extending into the retrosternal area, which cannot be clearly differentiated from the surrounding tissues. Yellow arrow: lymphadenopathies.

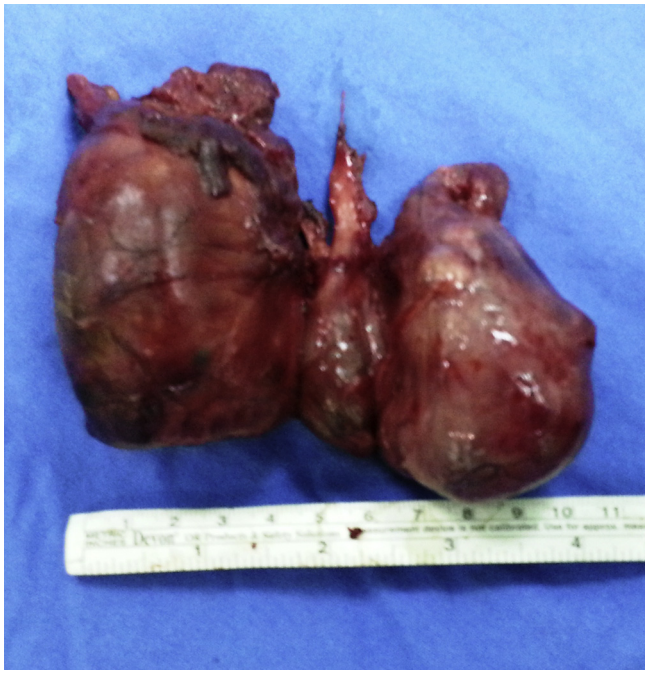

Fig. 2. The image of the total thyroidectomy specimen of the patient.

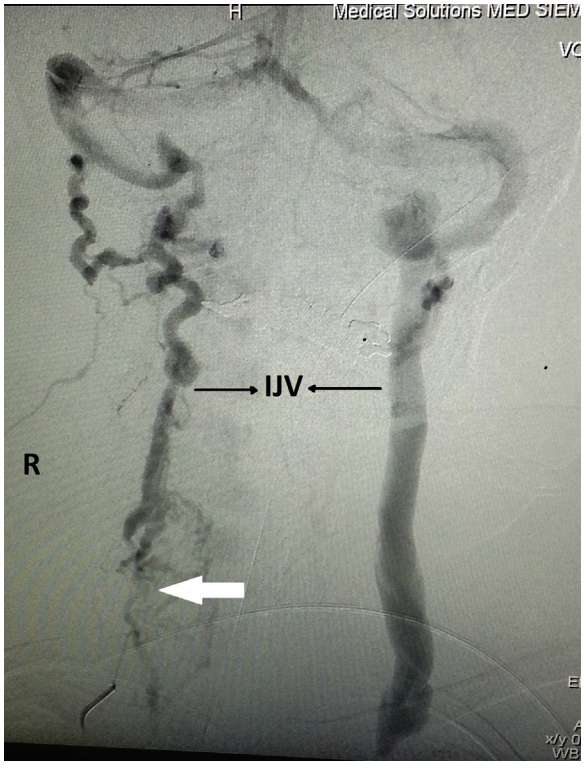

Fig. 3. The venography image of the patient performed after the development of the third hematoma. The right IJV is observed to be obliterated in inferior parts (r: right; white arrow: the obliterated area of the IJV).

contained micro-calcifications without clearly delineated borders, separating it from parenchyma. There were also multiple nodules featuring similar characteristics on the left lobe. Moreover, levels 2 and 3 of the right neck, as well as multiple lymphadenopathies (the largest of which was 14 × 17 × 24 mm in dimension) exhibiting the same characteristics as thyroid nodules were observed. Afterwards, FNAC was performed for both the right and left thyroid lobes, as well as for the lymphadenopathies on the neck region. The pathological results of the biopsies of both lobes and neck levels 2 and 3 were reported to be "compatible with thyroid anaplastic carcinoma". However, a widespread hematoma had developed on the right side of the patient's neck. The hematoma of the patient, who had a normal bleeding profile, as well as the patient's bleeding time and the thrombocyte functions regressed upon follow up. At that time, computed neck tomography (CT) was performed, and bilaterally increased heterogenecity featuring calcifications on the thyroid gland, an increase in the dimensions of the right sternocleidomastoid muscle (SCM), and multiple lymph nodes (the largest of which had a diameter of 3 cm) at the bilateral neck levels were reported. Moreover, increased densities were also found on the right supraclavicular adipose areas and on the skin due to edema. An increase in soft tissue density narrowing in the right paralaryngeal area and depletion of the larynx to the left were observed (Fig. 1). Upon physical examination, bilateral painless diffuse stiffness was palpated across the entire neck region. On endoscopic examination, the larynx was edematous and deviated to the left side with right vocal cord paralysis. Regarding his medical history, it was learned that he had a multinodular goiter for 10 years. In biochemical examination, thyroid stimulating hormone (1.63 uIU/mL), freeT_3 (3.26 pg/mL), freeT_4 (1.13 ng/dL), anti-thyroperoxidase antibody (5.04 IU/mL) and calcitonin (1.05 pg/mL) were normal but thyroglobulin antibody was high (619.4 IU/mL). Thorax CT revealed multiple nodules featuring a metastatic appearance in both lungs. Lung fiberoptic bronchoscopy revealed active hemorrhage originating from the right upper main bronchi, and brush and lavage samples were obtained. Upon pathological evaluation of these samples, malignity findings were not present. Afterwards, the patient was evaluated by our tumor board. To overcome respiratory deficiencies and to obtain an actual diagnosis, it was decided that surgery would be performed (a total thyroidectomy, if possible), versus biopsy and tracheotomy.

The operation began with a preoperative modified Kocher incision of 10 cm in length, but it was observed that the strap muscles were quite coherent with the thyroid. The results of the frozen section obtained from this region were "compatible with anaplastic carcinoma". Later, the operation was continued with inclusion of the strep muscles to the surgery. Although left lobe dissection was uneventful, during the right lobe dissection, the thyroid was